Middle ear malignant happened after lacrimal sac malignant

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Abstract
This paper reports a case of squamous cell carcinoma of middle ear that appeared after lacrimal sac malignant with an unusual clinical course.

Case report
A 57-year-old female patient was admitted to our hospital 5 years ago, with recurrent right side epistaxis and epiphora who was diagnosed with squamous cell carcinoma of lacrimal sac in our hospital, after surgery and the subsequent treatments of chemotherapy and radiotherapy, the patient had a good recovery. However, approximately 30 months later, the patient started to have symptom of purulent with blood discharge from the right ear, gradual hearing loss and tinnitus. Pathological examination revealed the diagnosis of squamous cell carcinoma of the middle ear (SCCME). Lacrimal sac is a rare site of squamous cell carcinoma, and SCCME is also rare, SCCME happens after lacrimal sac malignant has been never described in the literatures.

Introduction
There has been no known reported case of aural metastasis from lacrimal sac malignant tumors, lacrimal sac tumors are uncommon tumors with approximately only 300 cases described in the literature [1]. The malignancy rate of all lacrimal sac tumors is 5% to 75%. The most common malignant tumors are squamous cell carcinoma [1,2], 50% of malignant of the lacrimal sac recur and of the mortality rate is more than 50% [3]. The risk factors of which include human papilloma virus (HPV) and human immunodeficiency virus (HIV). HPV has been implicated in the etiology of the lacrimal sac neoplasia [4]. Squamous cell carcinoma occurs in middle ear (SCCME) is also rare, only accounts for 1.5% of temporal bone malignant tumors [5]. Currently, the etiological factors of SCCME are unknown, but possible causes are chronic otitis media and HPV infection [6,7]. The manifestations of SCCME are atypical in early stage and similar to chronic supportive otitis media easy to be misdiagnosed. Surgery together with radiotherapy is the main treatment approach to middle ear malignancy [8,9]. Here we report the original and first case of a patient with squamous cell carcinoma of lacrimal sac that might distant metastasis to the middle ear.

Case history
A 57-year-old female patient was admitted to our hospital 5 years ago, with recurrent right side epistaxis and epiphora in the past two years. Nasal endoscopy showed a cauliflower-like granulation tissue protruded from the inferior nasal meatus (Figure1a). A biopsy from this area showed a diagnosis of non-keratoniogizing squamous cell carcinoma.

The eye movement and visual acuity were not disturbed. Palpation of the neck showed regional lymph nodes were unremarkable. A computed tomography (CT) scan of the head and neck showed soft tissue density lesions in right lacrimal sac and inferior nasal meatus did not show any evidence of bony erosion (Figure1b), and chest CT scan did not disclose any abnormalities. After discussion at the multidisciplinary consultation meeting, surgery for removal of lesions in lacrimal sac and right inferior nasal meatus was decided. The patient underwent medial maxillectomy and dacryocystectomy through a lateral rhinotomy approach and surgical margins were all negative. Intraoperative pathologic biopsy from lacrimal sac tumor revealed squamous cell carcinoma in situ (Figure2a). Immunohistochemistry staining of CK was positive and performed for HPV marker. In situ hybridization was positive for high-risk HPV strains 16, 18 and negative for low-risk HPV strains 6 and 11. The patient was eventually diagnosed as squamous cell carcinoma of the lacrimal sac. Review of the magnetic resonance imaging scan for the surgical site taken soon after the surgical showed no evidence of local recurrence (Figure1c). The patient received postoperative chemotherapy and radiotherapy. No recurrence or metastasis was observed at the 12-month follow-up.

Key words: lacrimal sac, middle ear, squamous cell carcinoma, metastasis

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characteristics: the lacrimal sac is a rare site of squamous cell carcinoma, and SCCME is also rare. SCCME appears after lacrimal sac malignant has been never described in the literatures. In our case, the patient was diagnosed with chronic purulent otitis media before operation. Although infection of HPV is also the possible cause of SCCME, immunohistochemistry analysis of middle ear tumor tissue showed negative for all of high-risk HPV strains 16, 18 and low-risk HPV strains 6 and 11. Chronic otitis media is one of the etiological factors in SCCME. But in our case, the patient has no history of chronic otitis media. In the process of diagnoses, the results of preoperative CT shown for benign lesions. So she was mistaken as chronic supportive otitis media at first. But diagnosis of SCCME was confirmed by intraoperative frozen and postoperative pathology, which consistent with her history of lacrimal sac carcinoma pathology with the same side. In addition, although ear symptoms appeared 30 months after medial maxillectomy and dacryocystectomy, it might take a long time from onset to clinical symptoms. So, we speculate the SCCME may arise from the lacrimal sac carcinoma in a slow process. Several other possibilities may also explain the nature of the SCCME: First, it is possible that the SCCME arose de novo, independently in squamous cell carcinoma of lacrimal sacs, postoperative radiation may lead to the middle ear malignant tumor. A second possibility is that squamous cell carcinoma of lacrimal sacs could have led to seeding of the tumor on the ipsilateral middle ear. After receiving a radical mastoidectomy and radiotherapy, the patient have a good recovery within 6-month follow-up. Clinicians should take into account the possibility of distant metastasis in otitis media patients with a history of malignancy.

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